PDF hosted at the Radboud Repository of the Radboud University Nijmegen

The following full text is a publisher's version.

For additional information about this publication click this link.
http://hdl.handle.net/2066/21715

Please be advised that this information was generated on 2017-10-24 and may be subject to change.
Co-Occurrence of Plasma Cell Orificial Mucositis and Plasmoacanthoma

Report of a Case and Review of the Literature

Key Words
Mucositis
Plasma cell
Plasmoacanthoma

Abstract
Plasma cell orificial mucositis is a relatively rare condition which may occur on mucous membranes. Plasmoacanthoma is a verrucous tumor with plasmacytic infiltration and has been described as a separate disease entity. Such a tumor is supposed to occur without lesions which could be compatible with plasma cell orificial mucositis. The present case report documents the coexistence of both conditions in one single patient. This observation suggests that both conditions are part of one single disease entity.

In 1952, Zoon [1] described a predominantly plasmacytic infiltration of the mucosal membrane of the glans penis. This condition was designated as 'balanoposthite chronique circonscrite bénigne à plasmocytes'. Several authors have confirmed the existence of the chronic circumscribed benign balanoposthitis with plasma cells and have reported that other mucosal membranes might display the same pathological condition: the vulva, the lips and mucosal membranes of the oral cavity [2–16]. This condition, therefore, is best designated as 'plasma cell orificial mucositis' [16].

In 1962, Ferreira-Marques [17] described a patient with a tumor of the upper lip with verrucous aspect. The histopathological appearance of the tumor was characterized by hyperkeratosis, acanthosis and a predominantly plasmacytic infiltrate. The authors hypothesized that this condition represents a disease entity. In 1965, Ramos and Silva [18] reported a virtually identical case. No signs of plasma cell orificial mucositis were observed in both patients. Therefore, it has been suggested that plasma cell orificial mucositis and plasmoacanthoma are separate conditions.

In the present case, however, the patient was suffering both from plasma cell orificial mucositis and plasmoacanthoma. The nosological independency of both conditions is discussed in the light of the present case report and the available data in the literature.

Case Report

An 80-year-old woman presented with a painful condition of her lips. Two years before presentation at our department, the patient had noticed for the first time some redness and some scaling of the lips. There was no previous history of excessive exposure to the sun or signs of sun-damaged skin. The patient did not indicate a positive history of atopy. During the last months before presentation, the condition had worsened considerably and the patient complained of soreness of the lips. The patient never experienced an itchy sensation of the lips. The patient was not taking chewing gum, did not apply lipsticks and had not the habit of lip-biting and lip-licking. In the previous 2 years, the lips had been treated with Efudix cream, fusidine ointment and unguentum cetylicum without any success. Some relief from her complaints was reached following the application of the topical corticosteroid clobetasone butyrate in ointment (Emovate ointment).
Fig. 2. A verrucous tumor at the pullatum mole near the vulva.

Fig. 3. The histopathological appearance of a biopsy taken from the lower lip. A pronounced papillary epithelial proliferation is observed in the dermis. The epidermis is acanthotic and hyperkeratotic. Parakeratotic and parakeratotic cells are prominent.

Fig. 4. The histopathological appearance of a biopsy taken from the tumor of the soft palate (a). Note the striking acanthosis and papillomatosis in the dermis. The predominant cell type is composed of plasma cells and mononuclear cells.

Fig. 1. Swelling and erythema of the lips with fissuring.
The previous history revealed total hip replacement, venous leg ulceration and heart failure. For these reasons the patient had been treated with furosemide 40 mg once daily, nitrazepam 5 mg once daily, hydrochlorothiazide 25 mg three times per day and piracetam 800 mg three times per day.

At presentation an erythematous swelling of the upper and lower lip was observed. The swelling was complicated by deep fissuring (fig. 1). A slight to moderate lichenification was observed at the outer surface of the lips. No formation of vesicles had occurred. An indolent soft verrucous tumor was present at the palatum molle adjacent to the surface of the lips. No formation of vesicles had occurred. An indolent lip was observed. The swelling was complicated by deep fissuring. Blood investigations (clinical chemistry and hematology) were normal apart from a slightly increased sedimentation rate of 36 mm in the first hour. The Treponema pallidum hemagglutination test was negative.

Histopathological investigation of a biopsy taken from the lower lip revealed abnormalities of the dermal and epidermal compartment (fig. 3). In the dermis, a predominantly plasmacytic infiltrate was observed which partly penetrated into the epidermis. The plasma cells were not atypical. The epidermis was partly acanthotic and demonstrated spongiosis. The epidermis was invaded by several polymorphonuclear leukocytes. In the periodic acid-Schiff staining, no fungi were observed.

Histopathological investigation of a biopsy taken from the tumor of the palatum molle demonstrated an acanthotic epidermis and marked papillomatosis (fig. 4). The dermis was invaded by a dense inflammatory infiltrate composed mainly of plasma cells and T lymphocytes. The epidermis did not show parakeratosis, and no vacuolar degeneration of the upper malpighian layer was seen.

The condition of the lips was diagnosed as plasma cell orificial mucositis and the tumor at the palatum molle as plasmoacanthoma. As the tumor at the palatum was indolent, no treatment was initiated. The mucositis of the lips responded well to betamethasone dipropionate (Diprosone ointment, Essex).

Discussion

The changes of the lips have been diagnosed as plasma cell orificial mucositis. Numerous plasma cells were observed in the dermis and dermoepidermal interphase. Although some lichenification can be observed, the absence of vesiculation and itchy sensation, the negative responses of patch testing and the negative previous history of atopy do not substantiate the diagnoses of atopic cheilitis or allergic contact dermatitis. The lesions are not compatible with the diagnosis of actinic cheilitis, as the upper and lower lips are involved to the same extent and no signs of sun damage were observed.

The tumor of the palatum molle may resemble at first glance condylomata acuminata. However, the absence of hydropic degeneration in the upper layer of the stratum Malpighii and the absence of parakeratosis do not give any support for such a supposition. The lesion was diagnosed as plasmoacanthoma. The presence of a plasmacellular infiltrate in combination with the marked acanthosis and papillomatosis suggests that the lesion belongs to the same entity as the lesions reported by Ramos and Silva [18] and by Ferreira-Marques [17].

To the best of our knowledge, this is the first case report of the coexistence of plasma cell orificial mucositis and plasmoacanthoma. The simultaneous occurrence of both conditions in one single patient is compatible with one single disease entity.

The chronic circumscribed benign balanoposthitis with plasma cells is not characterized by epidermal acanthosis. In contrast, epidermal atrophy is usual in this condition. Plasma cell orificial mucositis may show epidermal atrophy [2, 3], slight acanthosis [16] and pronounced epidermal acanthosis with spongiosis and infiltration of polymorphonuclear leukocytes [12–15]. It is of pathogenetic importance that patients, who had lesions of the latter type with marked epidermal thickening and invasion of polymorphonuclear leukocytes, were habitual gum chewers [12–15]. In these patients, the mucositis improved or even disappeared after stopping gum chewing. It has been suggested by Goring et al. [19] that unspecified irritation such as mechanical trauma and candidiasis may induce plasmoacanthoma. Miller et al. [20] reported on mucositis with plasmocytic infiltration resulting from cinnamon allergy. It is remotely possible that plasma cell orificial mucositis is not one single disease entity. The mucositis in the present case is again characterized by epidermal acanthosis and invasion of granulocytes. The patient, however, was not using chewing gum, and no chronic irritant effect was found.

From a therapeutic point of view, a difference has been reported between atrophic and the mild acanthotic variants [16] and the marked acanthotic variant [21]. Again in the present case, suffering from the acanthotic variant, the cheilitis responded well to a potent topical corticosteroid.

The present case illustrates the coexistence of plasma cell orificial mucositis and plasmoacanthoma. The coexistence of both lesions suggests a common pathogenetic mechanism. The cell-biological explanation for the heterogeneity with respect to epidermal involvement remains to be further investigated.
References